Ectopic thyroid tissue within thyroglossal duct cyst as the only functioning thyroid tissue and it's management considerations

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ABSTRACT

Functional ectopic thyroid tissue may rarely be present within the walls of a thyroglossal duct cyst, and these may be the only functional thyroid tissue. We described one such case of ectopic thyroid within the thyroglossal duct cyst and its surgical management. A 12-year-old girl presented to us with a gradually increasing, painless midline neck mass that moved with swallowing and tongue protrusion. Ultrasonography showed a well-defined spongiform midline lesion measuring 3.2*1.4 cm in the upper neck. The thyroid scan showed focal increased tracer uptake in the midline upper neck corresponding to the clinically palpable swelling. A Sistrunk procedure was carried out, and the patient was started on thyroid hormone replacement.

Keywords: Ectopic thyroid, hypothyroidism, thyroglossal duct cyst, Sistrunk procedure.

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INTRODUCTION

Thyroglossal duct cysts (TGDC) are one of the commonest midline neck masses that arise as a cystic expansion of the remnant thyroglossal duct tract. These occur in approximately 7% of people and constitute over two-thirds of congenital neck masses. A TGDC usually presents as mobile, soft, painless neck swelling and can present at any location from the base of the tongue (foramen cecum) to the suprasternal notch.

Ectopic thyroid refers to the presence of functional thyroid tissue at places other than its normal anatomic location. It results from the failure of caudal migration of the thyroid, with an estimated incidence of approximately 1 in 200,000 people. The wall of TGDC is an uncommon location of ectopic thyroid tissue.

Herein, we describe a case of thyroglossal duct cyst with ectopic thyroid tissue in its walls and its surgical management in an adolescent girl.

CASE PRESENTATION

A 12-year-old girl was referred to our department with painless midline neck swelling that had been gradually increasing over the past two years. (Figure 1) Head and neck examination revealed a mass, approximately 3*2 cm^2 in the neck, anteroinferior to the hyoid bone. The mass was well-defined, cystic, firm, and non-tender on palpation and moved upward with swallowing and tongue protrusion. No other significant findings were found on the rest of the head and neck examination, including flexible

fiberoptic nasopharyngolaryngoscopy. Thyroid function test (TFT) showed subclinical hypothyroidism: TSH- 11.2 (reference range: 0.5-5), T3 and T4 within normal limits. Ultrasonography revealed a well-defined spongiform midline lesion measuring 3.2*1.4 cm in the upper neck, and the thyroid gland was not visualized at its normal location.

A technetium-99 m pertechnetate scan (Thyroid scan) showed focal increased tracer uptake in the midline upper neck. (Figure 2) Hybrid SPECT/CT localized tracer uptake to about a 2.5*1.4 cm^2 lesion in the midline in the upper neck, corresponding to the clinically palpable swelling. There was no definitive scan evidence of functioning thyroid tissue elsewhere in the neck and mediastinal regions.

The patient underwent a modified Sistrunk procedure under general anaesthesia. The entire 2*1.5 cm^2 of thyroglossal duct cyst was excised. (Figure 3) The patient had an uneventful postoperative course. Histopathology revealed multiple, variable sized thyroid follicles lined by cuboidal epithelium with abundant colloid production and scalloping suggestive of colloid nodule, confirming our diagnosis of thyroglossal duct cyst with ectopic thyroid. (Figure 4) She was started on thyroid hormone replacement therapy. On six weeks and three months follow-up, the TFT were within normal limits and she was doing well.

Figure 1: Neck examination revealing a midline neck mass

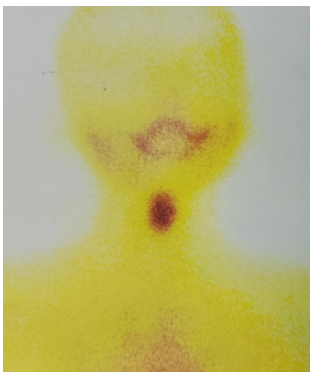

Figure 2: Thyroid scan showing uptake within the clinically palpable mass

Figure 3: Resected specimen of the thyroglossal duct cyst en masse

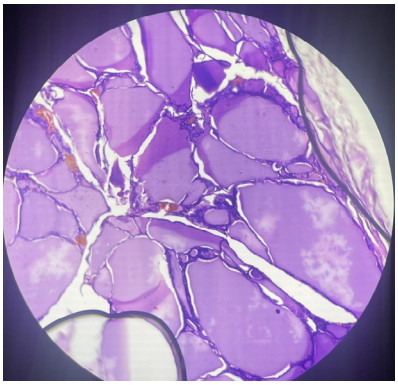

Figure 4: Variable-sized thyroid follicles filled with colloid

DISCUSSION

The thyroglossal cysts can occur at any location of normal thyroid migration, most commonly thyrohyoid, suprahyoid, suprasternal, and intra-lingual.[4] Most cases present in childhood or adolescence. Males and females are equally affected. The initial clinical presentation in most cases is an anterior neck swelling that moves with deglutition and tongue protrusion. Infection of the TGDC is a common presentation in children. Obstructive features/ features of mechanical impairment, such as dysphagia, hoarseness, and globus, are common features in adults.[5] Diagnosis is confirmed by imaging or fine-needle aspiration/ biopsy.

Ectopic thyroid tissue may reside at any location corresponding to its embryologic path of descent. Among ectopic thyroid glands, most are lingual (90%), and the rest occur at other sites.[6] Most thyroid ectopias manifest as simple TGDCs in conjunction with a normally developed thyroid gland in its usual thyroid cervical bed.[7] However, in our case, there was an absence of any thyroid tissue in its normal anatomical location, and all of the functional thyroid tissue was present within the wall of the TGDC, as evidenced by the thyroid scan.

Two-thirds of ectopic thyroid tissue function normally, and one-third may present with hypothyroidism.[8] Our patient

had subclinical hypothyroidism. The normal thyroid tissue reacts to increased demand by producing more thyroxine without abnormal enlargement. It is thought that ectopic thyroid tissue is dysgenetic and may enlarge because of stimulation by thyrotropin during a period of metabolic demand, like puberty, as was in our case, in which the cyst wall had the only functional thyroid tissue that enlarged during the pubertal age.

The management of TGDC depends on various factors, such as the presence of active infection or inflammation and surgical suitability. Infected TGDC should be treated initially with broad-spectrum antibiotics.[9] The definitive management consists of Sistrunk operation in suitable patients to prevent recurrent infection and to exclude cancer.[10] In our patient, the only functional thyroid tissue was within the TGDC, which was removed by modified Sistrunk procedure, and thus, lifelong thyroid hormone replacement therapy was given after thorough patient counselling.

CONCLUSIONS

Ectopic thyroid tissue may rarely present in a thyroglossal duct cyst and may be the only functional thyroid tissue. Normal thyroid tissue should be actively searched for before proceeding to surgery, as it influences management. Surgical excision remains the safe and effective procedure for TGDCs, but lifelong thyroid hormone replacement is needed if these contain the only functional thyroid tissue. Patient counselling is important in this regard.

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AUTHORS' CONTRIBUTIONS

BS contributed to concept, design, manuscript drafting and patient management. SK, AG, AA contributed to manuscript drafting, literature review and referencing. AD and BSH contributed to literature review, figure preparation and manuscript preparation. All authors read and approved the manuscript.

PATIENT CONSENT

The patient has provided informed consent for the publication of the case.

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